Recent Surgical Outcomes of Ebstein's Anomaly: A Focus on Changes in Outcomes Due to Cone Repair

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Abstract

Background: Ebstein's anomaly exhibits a wide variety of clinical features, and therefore, proposing a standardized treatment for it is difficult. This study was conducted to determine whether Cone repair, which has been implemented in our hospital since 2008, is more effective than conventional repair.

Methods: We retrospectively analyzed the clinical information of patients with Ebstein's anomaly who were followed-up at the hospital from 2000 to 2019. A total of 61 patients who had undergone tricuspid valve repair after 2000 were divided into the conventional and Cone repair groups and their clinical outcomes were compared.

Results: Of the 170 patients, 82 (48.2%) patients received surgical treatment for the tricuspid valve, whereas 75 patients received only medical treatment. The median follow-up duration was 5.89 years. After surgery, tricuspid valve regurgitation decreased and aortic stroke volume increased in both the Cone and conventional repair groups. In the Cone repair group, no mortality and postoperative complete atrioventricular block occurred and significantly fewer cases of moderate to severe tricuspid valve regurgitation were noted after surgery compared with the conventional repair group.

Conclusions: Cone repair is thought to be a method with less mortality and less occurrence of complete atrioventricular block than conventional repair.

Background

Ebstein's anomaly, a rare congenital heart disease, is characterized by the downward displacement of tricuspid valve (TV) leaflets into the right ventricle.[1,2] In addition, it is a disease that exhibits a wide variety of clinical manifestations because it is determined by various factors such as the degree of displacement of the TV, concomitant cardiac malformation, right ventricular (RV) dysfunction, arrhythmia, and left ventricular (LV) dysfunction. Although the downward displacement of the TV is commonly considered severe because the symptoms appear at a young age, the degree of RV dysfunction is severe with relatively mild symptoms in adults. Therefore, determining the optimal time for surgery remains controversial in clinical practice. Hesitation regarding the surgical treatment for Ebstein's anomaly has mainly been due to recurrent arrhythmia after surgery and the requirement for repetitive surgery. [3–6] However, with the introduction of the cone reconstruction technique in 2007 by da Silva and colleagues, [7] more anatomic repair has become possible compared with that with the previous Carpentier technique. Ebstein's anomaly repair with Cone reconstruction is being performed in our hospital since 2008, and this study was conducted to determine whether Cone reconstruction results in better clinical outcomes than conventional repair.

Methods

Study Populations

The medical records of patients diagnosed with Ebstein's anomaly who were treated and followed-up at our hospital from January 2000 to December 2019 were retrospectively analyzed for clinical information and treatment methods. Patients with Ebstein's anomaly and corrected transposition of the great arteries were excluded from the study.

Statistical Analysis and Ethics Approval

All continuous variables are expressed as means ± standard deviation. A compared mean test or the χ^2 test was performed to compare the differences in the paired variables between the Cone and conventional repair group. Statistical analyses were performed using SPSS version 18.0 for Windows (SPSS, Chicago, IL, USA). A *P* value of $\leq .05$ was considered statistically significant.

The Institutional Review Board of the Sejong General Hospital approved this retrospective study and waived the requirement for informed consent (approval number: 2020).

Results

This study included 170 patients, of which 108 (63.5%) were women. The number of cases diagnosed in the infantile period, childhood, and adulthood were 34 (20%), 63 (37.1%), and 73 (42.9%), respectively. The mean and median follow-up durations were 8.79 ± 8.26 years (range: 1 day to 33.91 years) and 5.89 years, respectively. The demographic characteristics of the patients are summarized in Table 1. A total of 144 (84.7%) patients out of 170 had New York Heart Association (NYHA) functional class I or II, indicating that relatively few patients had severe heart failure. At the first visit, the most frequent symptoms were dyspnea on exertion, followed by palpitation and chest discomfort. The most common associated congenital heart disease was atrial septal defect (ASD) or patent foramen ovale (PFO) in 92 (54.2%) patients.

Table 1
Demographic characteristics of Ebstein's anomaly patients

Variables	Number (%)
Sex	
Male	62 (36.5)
Female	108 (63.5)
Carpentier's classification	
A	63 (37.1)
B	67 (39.4)
C	34 (20.0)
D	1 (0.6)
N/A	5 (2.9)
Diagnosed Age	
Infant	34 (20.0)
Children	63 (37.1)
Adult	73 (42.9)
NYHA Functional class at diagnosis	
I	53 (31.2)
II	91 (53.5)
III	21 (12.4)
IV	5(2.9)
Symptoms at initial visit	
Dyspnea on exertion	91 (53.5)
Palpitation	60 (35.3)
Chest pain/chest discomfort	56 (32.9)
Cyanosis	28 (16.5)
Dizziness	18 (10.6)
Syncope/transient ischemic attack	14 (8.2)

Abbreviations: N/A, no available; ASD, atrial septal defect; NYHA, New York Heart Association; PAPVR, Partial anomalous pulmonary venous return; PDA, Patent ductus arteriosus; PFO, patent foramen ovale; VSD, ventricular septal defect

Variables	Number (%)
General edema/ascites	13 (7.6)
Combined congenital heart defect	
ASD or PFO	92 (54.1)
VSD	12 (7.1)
Pulmonary valve atresia or PS	12 (7.1)
PDA	8 (4.7)
PAPVR	1 (0.6)

Abbreviations: N/A, no available; ASD, atrial septal defect; NYHA, New York Heart Association; PAPVR, Partial anomalous pulmonary venous return; PDA, Patent ductus arteriosus; PFO, patent foramen ovale; VSD, ventricular septal defect

Although patients experienced several types of arrhythmias before the treatment of Ebstein's anomaly, only 60 (35.3%) patients experienced palpitations (Table 2). In addition, 17 patients had Wolff-Parkinson-White (WPW) syndrome, and arrhythmias originating from the atrium were more common than those originating from the ventricle. Moreover, determining whether some kinds of arrhythmia was an electrophysiological characteristic of Ebstein's anomaly or a secondary change due to enlargement of the right atrium was difficult. Furthermore, atrial flutter, atrial fibrillation, and sinus node dysfunction were observed.

Table 2
Types of arrhythmia identified in 85 patients prior to initiation of treatment

Types of arrhythmia	Number (%)
WPW syndrome with AVRT/NSAT	14 (8.2)
WPW syndrome with NSVT	1 (0.6)
WPW without arrhythmia event	2 (1.2)
1st degree AV block	6 (3.5)
Atrial flutter / Atrial fibrillation	17 (10)
AVRT/AVNRT/PSVT	14 (8.2)
Dual AV nodal pathway	1 (0.6)
AT	2 (1.2)
NSAT	7 (4.1)
Sinus node dysfunction	5 (2.9)
Ventricular tachycardia/ventricular fibrillation	3 (1.8)
NSVT	2 (1.2)
PAC and/or PVC	19 (11.2)
RBBB	3 (1.8)

Abbreviations: AT, atrial tachycardia; AV, atrioventricular; AVNRT, atrioventricular-nodal reentrant tachycardia; AVRT, atrioventricular reentrant tachycardia; NSAT, non-sustained atrial tachycardia; NSVT, non-sustained ventricular tachycardia; PAC, premature atrial contraction; PSVT, paroxysmal supraventricular tachycardia; PVC, premature ventricular contraction; RBBB, right bundle branch block; WPW, Wolff-Parkinson-White syndrome

The treatment methods of 170 patients according to the time period are presented in Table 3 and Fig. 1. A total of 75 (44.1%) patients opted for only medical follow-up without surgical treatment. Seven (4.1%) patients underwent tricuspid valve replacement (TVR) with tissue valve and 3 (1.8%) underwent TVR with mechanical valve, and 13 (7.6%) patients were treated only for other associated cardiac problems without TV repair. Since 1993, no TVR surgery has been performed using a mechanical valve. Cone repair was initiated in 2008. The trend of the change in the surgical method used at the Sejong General Hospital is presented in Fig. 2.

Table 3
Changes in the treatment method over time

Treatment methods	Patient's first visit year			Number (%)
	1986–1999 (n = 33)	2000–2007 (n = 62)	2008–2019 (n = 75)	
Medical treatment	4	27	44	75 (44.1)
TVP	19	28	25	72 (42.4)
Surgery or intervention without TV repair	5	4	4	13 (7.6)
TVR with tissue valve	3	2	2	7 (4.1)
TVR with mechanical valve	2	1	0	3 (1.8)
Abbreviations: TV, tricuspid valve; TVP, tricuspid valve plasty; TVR, tricuspid valve replacement.				

A total of 72 patients underwent tricuspid valve plasty (TVP). After 2000, 61 patients who underwent TVP were classified into the conventional and Cone repair groups and their surgical results were compared (Table 4). Compared with the conventional repair group, the Cone repair group had significantly greater aortic cross clamp (ACC) time and older age at operation. We compared the data of cardiac magnetic resonance imaging (MRI) performed within 6 months before and after surgery between the groups. No significant difference was noted between the two groups for the MRI data. After surgery, 6 (19.4%) patients of the conventional repair group had more than moderate degree of tricuspid valve regurgitation (TR). No post-operative complete atrioventricular (AV) block or operative mortality was noted in the Cone repair group. Five of the patients who underwent cone repair required reoperation, details of which are described in Table 5.

Table 4
Intergroup comparison of differences according to tricuspid valve repair technique since 2000

	Conventional repair group (n = 31)	Cone repair group (n = 30)	*P* value
Operation age (years)	19.54 ± 19.21	32.68 ± 19.11	.010*
FU duration (years)	11.73 ± 7.85	8.74 ± 19.11	.147
GOS ratio	1.33 ± 0.79	1.09 ± 0.55	.318
CPB time	277.13 ± 737.41	205.97 ± 56.55	.606
ACC time	83.57 ± 33.52	145.76 ± 51.39	0*
Cardiac MRI (preoperative/postoperative)	17/10	18/8	
Preoperative Aortic SV/BSA	31.80 ± 10.35	36.19 ± 6.23	.135
Preoperative Pulmonic SV/BSA	34.23 ± 19.22	36.03 ± 8.29	.726
Preoperative RVEDV/BSA	133.12 ± 67.41	175.17 ± 84.14	.114
Preoperative RVESV/BSA	69.35 ± 38.68	85.28 ± 47.89	.289
Preoperative RV EF (%)	48.88 ± 8.49	52.25 ± 6.27	.194
Preoperative LVEDV/BSA	57.82 ± 12.20	60.22 ± 10.54	.536
Preoperative LVESV/BSA	23.03 ± 7.72	22.33 ± 6.32	.772
Preoperative LV EF (%)	60.31 ± 10.10	63.64 ± 6.16	.253
Postoperative Aortic SV/BSA	41.61 ± 5.71	40.65 ± 6.98	.759
Postoperative Pulmonic SV/BSA	40.11 ± 7.19	34.84 ± 6.58	.125
Postoperative RVEDV/BSA	127.21 ± 30.17	105.63 ± 31.08	.156
Postoperative RVESV/BSA	68.53 ± 20.95	62.63 ± 24.58	.590
Postoperative RV EF (%)	46.58 ± 6.12	41.65 ± 8.31	.185
Postoperative LVEDV/BSA	71.97 ± 11.43	70.38 ± 7.85	.742
Postoperative LVESV/BSA	27.46 ± 7.78	26.88 ± 3.72	.848
Postoperative LV EF (%)	62.09 ± 5.57	61.20 ± 3.35	.681

* Statistically significant. Abbreviations: ACC, aortic cross clamp; AV, atrioventricular; BCPS, bi-directional Cavo-pulmonary shunt; BSA, body surface area; CPB, cardiopulmonary bypass; EF, ejection fraction; FU, fluorouracil; GOS, Great Ormond Street; LV, left ventricular; LVEDV, left ventricular end diastolic volume; LVESV, left ventricular end systolic volume; MRI, magnetic resonance imaging; RV, right ventricular; RVEDV, right ventricular end diastolic volume; RVESV, right ventricular end systolic volume; SV, stroke volume; TR, tricuspid valve regurgitation.

	Conventional repair group (n = 31)	Cone repair group (n = 30)	*P* value
Postoperative moderate to severe TR	6 (19.4%)	4 (13.3%)	.026*
BCPS add	5 (16.1%)	4 (13.3%)	1
mortality	3 (9.7%)	0	.081
Reoperation	10 (32.3%)	5 (16.7%)	.127
Surgical ablation	6 (19.4%)	8 (26.7%)	.554
Postoperative AV block	4 (12.9%)	0	.002*

* Statistically significant. Abbreviations: ACC, aortic cross clamp; AV, atrioventricular; BCPS, bi-directional Cavo-pulmonary shunt; BSA, body surface area; CPB, cardiopulmonary bypass; EF, ejection fraction; FU, fluorouracil; GOS, Great Ormond Street; LV, left ventricular; LVEDV, left ventricular end diastolic volume; LVESV, left ventricular end systolic volume; MRI, magnetic resonance imaging; RV, right ventricular; RVEDV, right ventricular end diastolic volume; RVESV, right ventricular end systolic volume; SV, stroke volume; TR, tricuspid valve regurgitation.

Table 5
Reoperation cases after Cone reconstruction for Ebstein's anomaly

Case number	Operation age	Preoperative state	Operation	Reoperation
45	54 years	RFCA performed for WPW syndrome, prolonged heart failure medication	Cone repair and ASD closure	1 year post-operation: TV septal and posterior leaflet detached 16 months post-operation: redo TVP and ring annuloplasty
109	1 year	Dyspnea and severe cyanosis, preoperative cardiac arrest	Cone repair	1 day post-operation: BCPS add and ECMO support 7 years post-operation: redo TVP
135	12 days	Percutaneous PVP state, but sustained cyanosis and dyspnea	Cone repair, BT shunt add, ASD partial closure, thrombectomy of right SVC	26 days post-operation: RA thrombus removed, TV repair, and PV repair performed 4 months post-operation: atrial septectomy, BCPS add, shunt division
139	26 years	Dyspnea, chest pain, atrial flutter, NYHA functional class III	Cone repair with right side Maze	2 days post-operation: posterior leaflet detached, TVR with bioprosthetic valve performed
140	2.3 years	No significant symptom, NYHA functional class II	Cone repair with BCPS	39 months post-operation: VT event, Fontan operation performed

Abbreviations: ASD, atrial septal defect; BCPS, bi-directional Cavo-pulmonary shunt; BT, Blalock–Taussig; ECMO, extracorporeal membrane oxygenation; NYHA, New York Heart Association; PV, pulmonary valve; PVP, peripheral venous pressure; RA, right atrial; RFCA, radiofrequency catheter ablation; SVC, superior vena cava; TV, tricuspid valve; TVP, tricuspid valve plasty; TVR, tricuspid valve replacement; VT, ventricular tachycardia; WPW, Wolff-Parkinson-White.

The 10-, 20-, and 30-year survival rates of all patients with Ebstein's anomaly who underwent follow-up at our hospital were 92%, 87.6%, and 51.6%, respectively. The survival curves of patients according to the treatment method are presented in Fig. 3.

Discussion

Ebstein's anomaly is a rare disease that exhibits a wide variety of clinical features, making it difficult for clinicians to consistently accumulate experience.[8–11] Generally, neonatal Ebstein's anomaly is known to be hemodynamically unstable, difficult for surgical correction, and associated with high mortality.

However, in the case of non-severe Ebstein's anomaly, the diagnosis is often delayed because the symptoms perceived by the patient are unclear. In fact, 42.9% of the patients visiting our hospital were diagnosed with the disease only in adulthood, of which 84.7% of the patients had NYHA functional class I or II. As such, patients' symptoms are often not severe, and as can be seen in Table 3, there have been quite a lot of cases of medical treatment rather than active surgical correction. Another big reason for encouraging the judgment of such medical support is that the experience of follow-up observation with medical support was not bad in terms of survival compared with that of various active surgical treatments (Fig. 1). Until relatively recently, there have been concerns about whether surgical correction for Ebstein's anomaly has an advantage over medical support because of the increase in mortality and morbidity due to repeated reoperations after surgical repair. Indeed, the results of our hospital as well as other centers reveal that the 10-year survival of Ebstein's anomaly is more than 90%. [5, 9, 12, 13]

Many studies have reported the excellence of cone repair,[14–18] however, although it is performed in our institution from 2008, only 30 operations have been performed till date. A total of 5 patients required reoperation after cone repair; however, patients who underwent cone repair from 2014 did not require a reoperation (Table 5). Therefore, it can be considered that we have overcome the initial learning period relatively quickly. Remarkably, the proportion of patients with complete AV block occurrence and those with moderate or severe TR remaining after surgery were significantly less in the Cone repair group than in the conventional repair group. In addition, in the case of Cone repair, no mortality was reported (Table 4). Moreover, because the age of the Cone repair group patients was significantly higher at surgery, it may be thought that patients with a relatively mild form of Ebstein's anomaly were included. However, cone repair was not specifically avoided in patients of young age.

Before the start of treatment, a large number of patients (85 patients, 50%) had already experienced several types of arrhythmias (Table 2). Some patients experienced multiple types of arrhythmias. Of the patients whose arrhythmia was found before starting treatment, 27 patients did not show arrhythmias after treatment. However, among the patients whose arrhythmia was not clearly identified before treatment, 33 cases showed new arrhythmias after starting treatment. This suggests that the burden of arrhythmia is very high in the case of Ebstein's anomaly.[18] It is thought that arrhythmia may develop even after improving the hemodynamic state and that responding with active interest is necessary.

Studies have proved that Ebstein's anomaly is not a simple tricuspid valve abnormality and is associated with LV or left atrial dysfunction. [19, 20] However, it is not easy to present standardized indicators in clinical practice. It is known than hemodynamic indicators determined by cardiac MRI can be used as relatively objective indicators because there are several limitations in evaluating RV function and TR using echocardiography. Thus, it is difficult to generalize because it was not performed in all patients, but the results of cardiac MRI performed within 6 months and after surgery were reported for 17 patients of the conventional repair group (54.8%) and 18 patients of the Cone repair group (60%). However, no statistically significant difference was noted between the conventional and Cone repair group. After surgery, the degree of TR significantly decreased in both the groups, as well as the right ventricular end diastolic volume (RVEDV), right ventricular end systolic volume (RVESV), and RV ejection fraction (EF)

presented a decreasing tendency (Table 4). It seems reasonable to interpret these changes as changes associated with a decrease in TR amount rather than a significant decrease in RV function after surgery. In addition, postoperative aortic stroke volume (SV) increased after surgery in both the groups. This result can be interpreted as after surgery, the overall cardiac output increased. Based on these results, if no mortality is associated with surgery and the frequency of reoperation is significantly lower than that of TVR or conventional TV plasty, it can be expected that chronic low cardiac output can be more actively improved and chronic hepatic congestion or right heart failure can be reduced by Cone repair. In other words, if we actively advance the operation age for Ebstein's anomaly than the existing age, it can expected that the outcome of patients will be slightly improved.

Study Limitations

This was a single center study with a small number of patients. In addition, limited data were analyzed due to the retrospective nature of the study.

Conclusions

Among the surgical treatment methods for Ebstein's anomaly, TVR increases the patient's discomfort and morbidity due to repeated reoperations. However, with surgical treatment, it is possible to reduce the TR amount and increase the cardiac output. TVP with Cone repair has a lesser risk of reoperation, mortality, and complete AV block than conventional repair.

List Of Abbreviations

ACC aortic cross clamp

ASD atrial septal defect

AV atrioventricular

BSA body surface area

CPB cardiopulmonary bypass

EF ejection fraction

GOS Great Ormond Street

LV left ventricular

LVEDV left ventricular end diastolic volume

LVESV left ventricular end systolic volume

MRI magnetic resonance imaging

NYHA New York Heart Association

PFO patent foramen ovale

RV right ventricular

RVEDV right ventricular end diastolic volume

RVESV right ventricular end systolic volume

SD standard deviation

SV stroke volume

TR tricuspid valve regurgitation

TV tricuspid valve

TVP tricuspid valve plasty

TVR tricuspid valve replacement

WPW Wolff-Parkinson-White

Declarations

Ethical approval and consent to participate

The Institutional Review Board of the Sejong General Hospital approved this retrospective study and waived the requirement for informed consent (approval number: 2020).

Consent for publication

Not applicable.

Availability of supporting data

All data generated or analyzed during this study are included in this published article.

Competing interests

The authors declare that they have no competing interests.

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Author's contributions

Eun-Young Choi, Jung Yoon Kim, and Eung Re Kim performed collected data and statistical analysis. Eun-Young Choi wrote this paper. Eun Sun Kim and Seong-Ho Kim supervised the overall completion of the project. Jae Hong Lim and Chang-Ha Lee contributed to the preparation of the figures and tables. All authors read and approved the final manuscript.

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Conflict of Interest and Source of Funding

Authors have nothing to disclose with regard to commercial support. We have no conflicts of interest.

Institutional Review Board Approval

This study was approved by the Institutional Review Board of the Sejong General Hospital (approval number: 2020; approval date: July 15, 2020)

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Figures

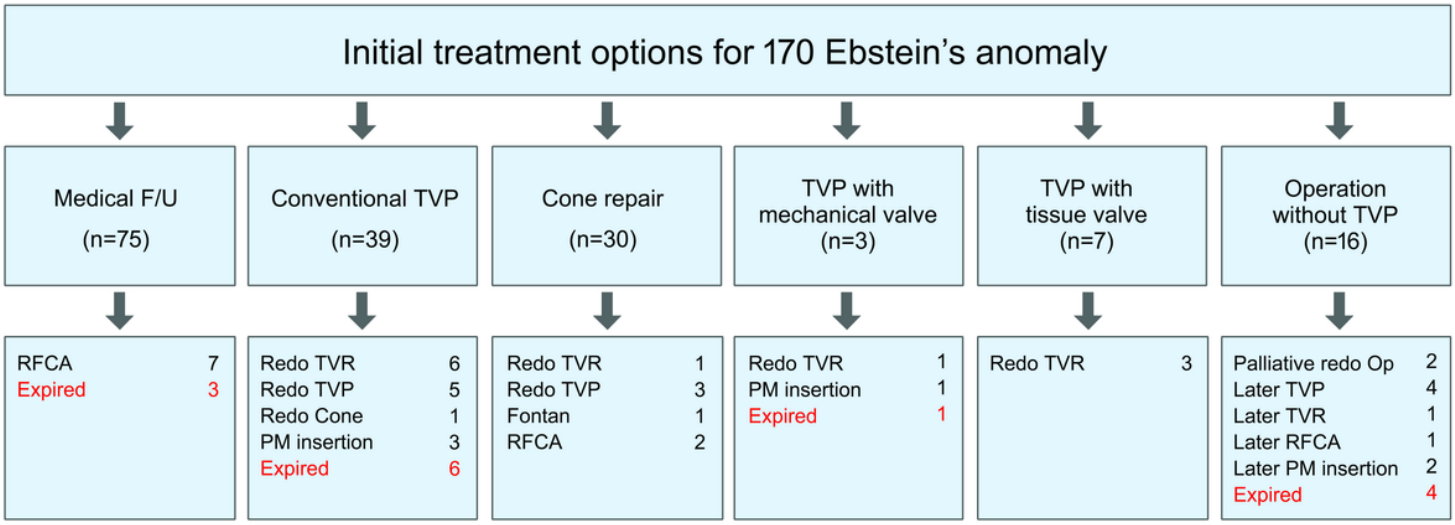

Figure 1

Initial treatment methods and clinical outcomes of 170 Ebstein's anomaly cases

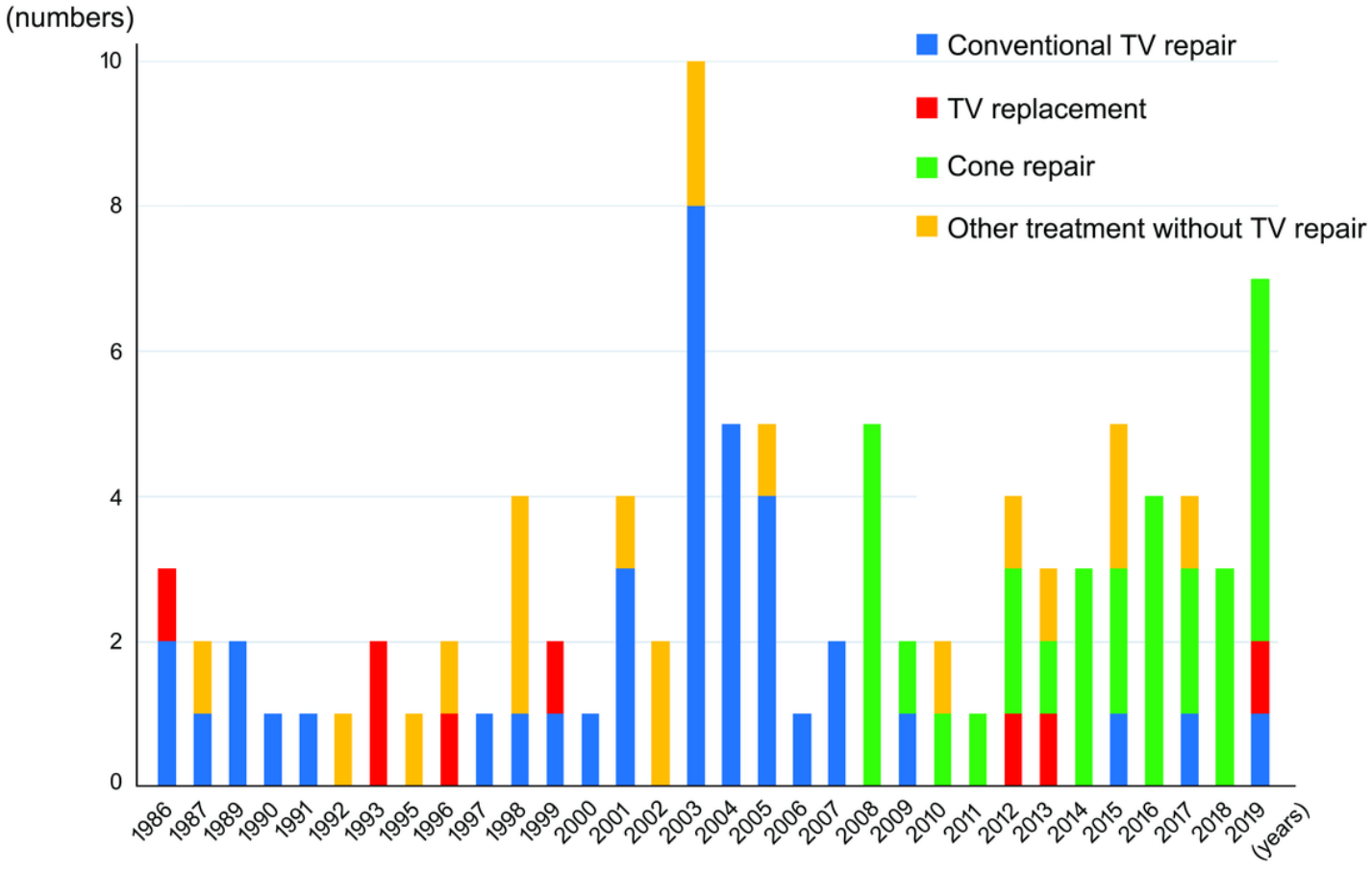

Figure 2

Changes in the surgical method by year

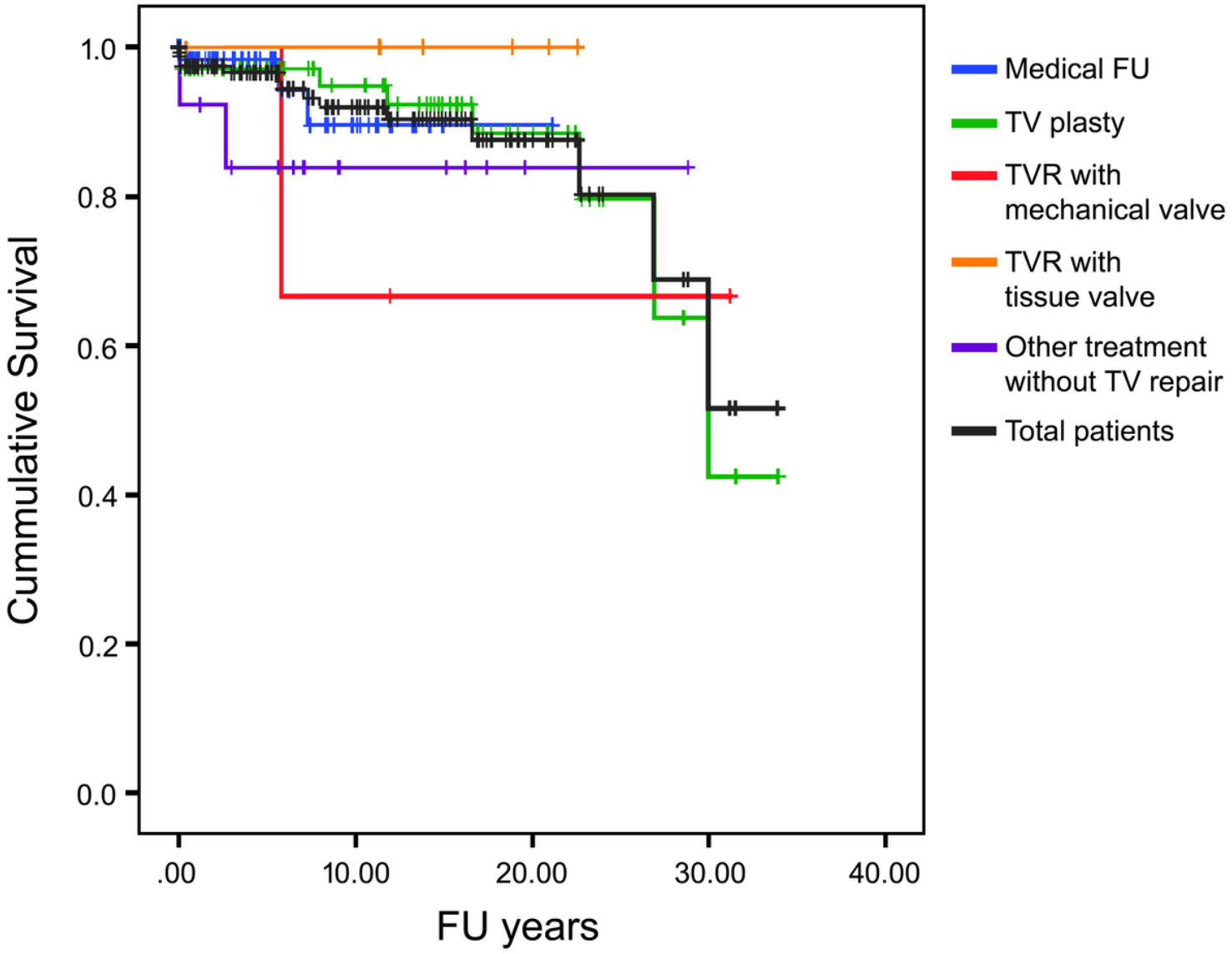

Figure 3

Comparison of survival rates according to treatment methods for Ebstein's anomaly